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MR. CALLUM WEMYSS (Orcid ID : 0000-0003-0107-0361)

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A case of idiopathic First Bite Syndrome responding to carbamazepine.

Callum Wemyss BDS (Hons) MFDS RCPS(Glasg)

Dental Core Trainee 3

Oral Surgery/Oral Medicine

Glasgow Dental Hospital and School

378 Sauchiehall Street

Glasgow

G2 3JZ

John Gibson PhD BDS MB ChB FRCP(Glasg) FDS(OM)RCPS(Glasg) FFDRCS(Irel)

Professor of Oral Medicine

Institute of Dentistry

University of Aberdeen

Cornhill Rd

Aberdeen

AB25 2ZR

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Kevin Ryan BDS MB BCh BAO FDS(OM)RCPS(Glasg)

Consultant in Oral Medicine (Clinical Lead)

Honorary Senior Clinical Lecturer

Glasgow Dental Hospital and School

378 Sauchiehall Street

Glasgow

G2 3JZ

Correspondence to

Callum Wemyss

Glasgow Dental Hospital and School

378 Sauchiehall Street

Glasgow

G2 3JZ

Email: callumwemyss@nhs.net

Phone: 0141 211 9600

Abstract

First Bite Syndrome (FBS) is a painful condition which classically presents as severe pain in the pre-auricular region, initiated on the first bite of a meal. It is usually associated with a history of upper neck surgery or tumour of the parotid salivary gland or parapharyngeal space (PPS). Some propose FBS arises from damage to, or resection of, the cervical sympathetic trunk resulting in loss of sympathetic innervation to the parotid salivary gland. Idiopathic FBS has been reported only a handful of times in the literature. We present the case of a 51-year-old male, urgently referred to the Department of Oral Medicine, with a three-week history of severe, sharp, unilateral pain from the right angle of mandible, intensity described as '20/10', occurring only on the first bite of eating. This would diminish over two minutes. Clinical examination revealed tenderness over the right condylar neck and angle of mandible only. FBS was suspected and the patient immediately started on carbamazepine. An urgent MRI displayed no abnormalities of the TMJ, PPS, trigeminal nerve or parotid gland. Restorative Dentistry review confirmed no odontogenic/occlusal cause and on review, four months later the patient reported complete resolution of symptoms on 200mg of

carbamazepine three times daily. This case highlights the importance of taking a thorough pain history. Patients with symptoms of FBS and no history of surgery to the neck should have an MRI to rule out parotid gland or PPS tumour. Carbamazepine can be successful in treating idiopathic FBS and can resolve symptoms completely.

Keywords: First bite syndrome, parotid gland, carbamazepine, head and neck surgery, temporomandibular pain.

Introduction

First Bite syndrome, first described by Netterville in 1998, is severe parotid pain elicited on the first bite of a meal without any prodromal symptoms.¹ This is not to be confused with the definition used by Haubrich in 1986 who used the term to describe a completely different collection of symptoms including an inability to swallow with retrosternal pain on the first bite of meal.² FBS usually diminishes seconds or minutes after the first bite of a meal, can deter patients from eating and has been reported to affect quality of life (QoL).³ It is thought to be accentuated by particular gustatory stimuli (acids, sour, spicy or sweet foods or drinks), salivation and some even say the thought of food.^{4,5}

It is typically seen in patients who have had surgery to the neck or a tumour of the parotid salivary gland or parapharyngeal space (PPS). Netterville's 1998 case series presented 49 patients who were treated with surgery for vagal paragangliomas.¹ First bite syndrome was reported postoperatively in eight of the nine patients who had sympathetic trunk resection. Since then, FBS has also been implicated post-operatively in cases of cervical lymph node dissection,⁶ deep lobe parotidectomy,⁷ TMJ joint replacement,⁸ bimaxillary osteotomy,⁹ resection of the stylohyoid ligament in Eagle's Syndrome,¹⁰ carotid bifurcation and/or internal carotid artery surgery such as carotid endarterectomy or ligation.¹¹ In a case series by Kawashima FBS was reported post-operatively in 9 of 29 patient who underwent resection of parapharyngeal tumours.¹² Following surgery it usually takes a few weeks but can sometimes take several months for symptoms to develop. In addition, there has been one recent case of Eagle's syndrome presenting with symptoms of FBS pre-operatively.¹³

Tumours reported to have caused symptoms of first bite syndrome include tumours of the parapharyngeal space (synovial sarcoma),¹⁴ deep poles of the parotid (adenoid cystic carcinoma, squamous cell carcinoma and mucoepidermoid carcinoma),¹⁵⁻¹⁷ and submandibular gland (adenoid cystic carcinoma).¹⁸ Caution must therefore be exercised with patients experiencing symptoms of FBS. Guss et al reported an adenoid cystic carcinoma of the submandibular gland which was not clinically or radiographically apparent until 18 months after initial presentation of FBS symptoms.¹⁸

More recently Maheer et al described a male, with no surgical history, diagnosed with FBS nine years previously who had a CT scan six years later which revealed a mass consistent with a lipoma in the right parotid region.¹⁶ After no improvement in pain with various drugs and an enlarging mass on the right face he was diagnosed with a squamous cell carcinoma of the parotid. It is unlikely he had the malignancy for nine years and the exact aetiology was concluded as uncertain. The only report of a benign tumour presenting as itself as FBS is an Osteoid Osteoma of the temporal bone.¹⁹

There have been a number of post-operative cases which have also been associated with Horner's syndrome which can also result from damage to the sympathetic trunk. Frey's, Harlequin's and Pour Tour du Petit are other examples of different symptom complexes resulting from injury to sympathetic innervation.²⁰

To date there have only been four published reports (one case series and three case reports) on patients with symptoms of FBS with no history of surgery and no detectable tumour on Magnetic Resonance Imaging (MRI).²¹⁻²⁴

Case Report

We present the case of a 51-year-old male, with no history of head or neck surgery, urgently referred to the Oral Medicine Department with 'jaw pain'. The patient presented with a three week history of severe, sharp, unilateral pain from the right angle of mandible, intensity described as '20/10' on a visual analogue scale (VAS), occurring only on the first bite of eating a meal. This pain would always diminish over two minutes and after it had subsided he could finish the meal without any further pain. The pain radiated to the ear and was accentuated by sweet foods. Prior to the referral the patient had been managed by his dentist for a temporomandibular disorder including conservative advice, analgesia, hard acrylic splint and jaw exercises.

Medically the patient had ischaemic heart disease and had a myocardial infarction 11 years ago. In addition, he had type II diabetes which was diet controlled. His most recent HbA1c was 72. He had no habits, history of trauma to the face and was not aware of clenching or grinding his teeth. He wore an upper cobalt chrome partial denture.

Clinical examination revealed tenderness over the right condylar neck and angle of mandible only. This was consistent with the position of the right parotid gland. There was no clicking, locking or crepitus of the temporomandibular joints (TMJs) and the joints were not tender to palpate. Mouth opening was unaffected. Furthermore, there were no enlarged salivary glands and no palpable cervical lymphadenopathy. Intraorally, the patient had a number of missing teeth and significantly

none posterior to the LR5 (lower right first premolar) on the lower arch. The UR6 (upper right first molar) and UR7 (upper right second molar) had large amalgam restorations. None of the teeth provided positive responses to ethyl chloride (cold testing) but gave normal range positive responses to the electric pulp test. This suggested, but did not confirm, both molars were vital.

An orthopantomogram (OPT) was taken (figure one) to rule out any odontogenic cause, or indeed, any other gross pathology. The OPT showed a retained root in the upper right quadrant, possible caries in the UR6 and LL4 (lower left first premolar) and severe bone loss due to periodontitis but nothing to account for the patient's severe pain.

FBS was suspected and the patient immediately started on carbamazepine 100mg once a day and his general medical practitioner asked to increase this as per response. An urgent MRI was carried out displaying some minor age-related changes but no abnormalities of the TMJ, PPS, parotid and trigeminal nerve.

The patient was reviewed after a month reporting an improvement on 100mg carbamazepine in the morning and 200mg at night. At this point a dedicated TMJ MRI sequence was carried out this showed some stretching of the disc on both sides but normal relocation and no other abnormalities. Restorative Dentistry review confirmed no odontogenic or occlusal cause to the patient's pain and on review, in the Oral Medicine Department, four months later the patient reported complete resolution of symptoms on 200mg of carbamazepine three times daily. The patient will be closely followed up with the plan to eventually taper off the carbamazepine.

Figure 1 OPT which includes the jaws, dentition, maxillary sinuses and TMJ.

Discussion

Netterville suggested that this syndrome is secondary to the loss of sympathetic innervation to the parotid gland.¹ This would result in denervation hypersensitivity of the sympathetic receptors that control the myoepithelial cells in the parotid gland. With oral intake, the secreted parasympathetic neurotransmitter cross-stimulates the sympathetic receptors, causing an autonomic imbalance. This leads to supramaximal contraction of the myoepithelial cells and pain on the parotid region on the first bite. Sympathetic innervation should not be damaged in idiopathic cases of FBS therefore this could suggest a different pathophysiology. There have been a number of cases following surgery involving ablation of the sympathetic trunk that did not develop first bite syndrome. Conversely, a deep lobe parotidectomy, due to a pleomorphic adenoma, where there was no reported damage to sympathetic structures and no ligation of the external carotid artery developed FBS.¹² These both

complicate the theory and support the fact that the pathophysiology of FBS is still not well understood.

Chiba et al reported 14 patients with idiopathic FBS who presented to an orofacial clinic over three years.²¹ Of these patients, nine had poorly controlled diabetes. Postprandial pain (pain after mealtime) was also suffered in six of the nine patients. Postprandial pain has not been reported in any previous case of post-operative or tumour-related FBS. Chiba et al also postulates that the pathophysiology is different and for that reason they refer to idiopathic first bite syndrome as Idiopathic Parotid Pain (IPP) in their paper. There have been a small number of idiopathic FBS cases which have been bilateral, again, hinting at a systemic rather than local mechanism in these cases. In the case, reported in this manuscript, the patient had poorly controlled type II diabetes but reported no postprandial pain. Diabetic neuropathy perhaps plays a role in idiopathic FBS but is unlikely to be the cause. Chiba et al highlighted that idiopathic FBS appeared to be more common in males and suggests that this could be due to the fact men more frequently develop diabetic neuropathy. Other than Chiba's case series there have only been three other cases of idiopathic FBS reported in the literature.²²⁻²⁴

There have been a number of suggestions that FBS, specifically post-operative FBS, will eventually resolve without any treatment over a period of months to years.²⁵ In a retrospective review of 53 patients undergoing surgery to the neck, 16 developed FBS postoperatively. Symptoms resolved in 11 of these patients over a period of months to two years without any treatment and the 5 remaining patients were still symptomatic at five years. Due to limited data and studies regarding idiopathic FBS it is unclear if symptoms would completely resolve without any treatment.

A number of different treatments have been used and suggested for the management of first bite syndrome. The most common treatment appears to be pharmacological management. Non-steroidal anti-inflammatories and codeine-based analgesia have proved ineffective in all cases. Other drugs used have included carbamazepine (with or without amitriptyline), pregabalin and neurotrophic agents.²⁰ In the idiopathic case, which we present, the patient's pain was significantly better at one month of carbamazepine and completely resolved by five months. Stoopler and Elmuradi reported an idiopathic case of FBS which responded to botulinum toxin (botox) injections into the parotid gland after not responding to 150mg oxcarbamazepine twice a day and 1800mg gabapentin daily.²² To date, botox has been reported effective, in reducing pain in all cases it has been used.^{3,26-29} Typically the effects last three-four months at which point patients require repeat injections, however, it has been suggested that due to the fact that symptoms of FBS can resolve with time it could be a useful temporising measure.²⁷ There have been no side effects reported such as facial paralysis or dry mouth associated with intraglandular injections of botox.

In non-idiopathic cases, dietary modification, such as asking the patient to stick to 'bland' foods, has been suggested as a successful treatment.¹ Chiba reported that one of the of the eight idiopathic FBS patients responded to dietary modification with the rest responding to pregabalin or neurotrophic agents.²¹

Stellate ganglion blocks have been used in another idiopathic case by Kurokawa et al to effect and acupuncture has been used to varying effect.^{23,30} Radiotherapy has also been cited as a successful treatment in patients who developed FBS following major oncologic neck surgery. This is perhaps due to damage to parotid parenchyma from the radiation. In a study of 499 patients who had surgery to the parotid, PPS or infratemporal fossa (ITF) between 1992-2010, 9.6% of patients developed FBS. Of the 499 patients, 47 of these patients had a history of head or neck radiotherapy and none went on to develop FBS.³¹

Surgery to remove tumours causing FBS have proven successful in resolving symptoms but surgery to treat first bite syndrome in post-operative cases, such as tympanic neurectomy, has proven unsuccessful and has certain risks attached.^{5,26} There has only been one reported case of resolution with laser tympanic plexus ablation, however the follow-up for this case was only two months.³²

Conclusions

This case highlights the importance of taking a thorough pain history. FBS should not be confused with temporomandibular pain which could lead to inappropriate management. Patients with symptoms of FBS and no history of surgery to the neck should have an MRI to rule out a parotid, PPS or ITF tumour. Diabetes could play a part in idiopathic cases of FBS and this should be further investigated. Carbamazepine can be successful in treating idiopathic FBS and can resolve symptoms completely.

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